

## Two Cases of Adventitial Cystic Disease of the External Iliac Vein

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**Introduction:** Adventitial cystic disease (ACD) affecting veins is a very rare condition.

**Report:** We report two cases of ACD affecting veins in female patients aged 50 and 32 years who underwent surgical excision.

**Discussion:** Venous ACD presents with deep venous thrombosis (DVT)-mimic symptoms resulting from venous compression by the cystic mass. Optimal treatment of venous ACD is complete surgical removal of the cystic wall and its contents; however, recurrent symptoms are often reported because of recurrence of the cystic mass.

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### INTRODUCTION

Adventitial cystic disease (ACD) more commonly affects an arterial segment, such as popliteal or iliac arteries. Venous ACD is very rare. From the first case of venous ACD reported in 1963, less than 40 cases have been reported in English literature.<sup>1,2</sup> We report two cases of venous ACD describing clinical features and treatment results. Ethical approval for the research was granted in institution.

### CASE REPORTS

#### Case 1

A 50-year-old female presented with gradual swelling of her left leg over 20 days. She denied any history of trauma or past deep vein thrombosis (DVT). Hypercoagulability study showed no abnormalities. Duplex showed a  $4.4 \times 2.7$  cm cyst compressing the left external iliac vein (EIV) with no DVT (Fig. 1A), and magnetic resonance imaging (MRI) showed high signal intensity lesions on T2-weighted image (Fig. 1B).

Surgical removal of the cyst was performed. The EIV and common femoral vein (CFV) were exposed through vertical incision crossing inguinal ligament. We found a  $4 \times 2$  cm, unilocular cyst with amber-colored-gelatinous material compressing the EIV (Fig. 1C). After evacuation of material and complete excision of the cystic wall, the EIV and CFV spontaneously recovered. There was no recurrence on follow-up until 12 months. Pathologic examination revealed intramural cystic wall with increased proteoglycan and a few fragmented elastic fibers with diagnosis of ACD (Fig. 1D).

#### Case 2

A 32-year-old female was referred to us with increasing swelling in her left leg. Seven years earlier, she had undergone cyst removal and open thrombectomy of the left EIV at another hospital under impression of cyst with acute DVT. She received anticoagulation therapy postoperatively for 12 months. The iliac DVT recurred and she underwent catheter-directed thrombolysis 3 months before referral to us. On laboratory tests, there were no abnormalities. Duplex and MRI revealed a multilocular cyst compressing the EIV with no DVT.

At operation, we found a multilocular cyst with fibrosis and stenosis of the EIV. After opening the cyst, amber-colored gelatinous material was evacuated. After removal of the cyst, we found EIV with stenosis and wall thickness but there was no thrombus in the EIV. We performed patch venoplasty with bovine pericardium. Postoperatively, we prescribed anticoagulation. Three months after the operation, we found the EIV at venoplasty site but a recurrent cyst in follow-up duplex. We again performed surgical resection of the recurred cyst, but we could not perform complete resection of the cyst.

### DISCUSSION

According to review of previous reports of 34 venous ACD patients, ACD often affected lower extremity veins including femoral (53%), iliac (26%), popliteal (9%) and great saphenous (9%) veins.<sup>1,2</sup> On the other hand, arterial ACD most commonly occurred in the popliteal artery, but was also reported in the iliac, femoral, and radial arteries.<sup>3</sup>

Clinically, venous ACD can be difficult to detect with physical examination. For definitive diagnosis, imaging studies such as duplex scans, computed tomography (CT) scans, or MRI are reported to be equally efficacious.<sup>4</sup> The number of unilocular cysts in past reports was similar to multilocular cyst venous ACD.<sup>1,2</sup>

For venous ACD, open surgical excision of the entire wall is the treatment of choice.<sup>1</sup> Percutaneous angioplasty and image-guided aspiration of cysts is known to be an

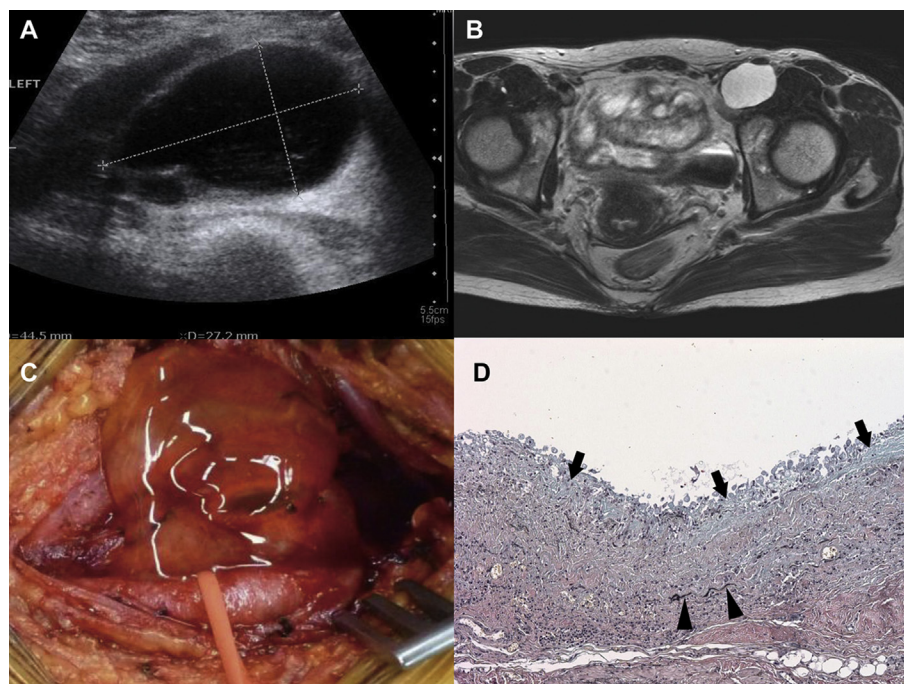
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**Figure 1.** Diagnostic imaging and operative findings of venous ACD in a 50-year-old woman. (A) A typical hypoechoic fluid-filled cyst with a posterior acoustic window on duplex ultrasonography. (B) MRI shows a high signal intensity lesion compressing the left external iliac vein on a T2-weighted image. (C) On opening the cyst, thick gelatinous material issued from the cystic cavity close to the external iliac vein in operative finding, and (D) pathologic examination of the surgical specimen revealed that the cystic wall is composed of fibrous tissue with increased proteoglycan (arrows) and a few fragmented elastic fibers (arrow heads). (MOVAT stain, original magnification  $\times 100$ ).

ineffective treatment because of high recurrence rates.<sup>1,4</sup> Percutaneous aspiration and injection of sclerosing agent have been reported only as a case report.<sup>5</sup> After surgical treatment, recurrences have been reported following simple evacuation of cystic material or incomplete excision of cystic wall.<sup>1,2</sup> Patients with venous occlusion may necessitate resection of the affected vein and reconstruction with interposition graft or patch angioplasty similar to arterial ACD.<sup>1,2</sup> Long-term results of surgical treatment are still unknown because of the rarity of venous ACD. But, it is important to necessitate accurate diagnosis and complete resection of venous ACD for prevention of recurrence.

In summary, venous ACD is a very rare disorder in differential diagnoses for leg swelling. Cystic lesion compressing the vein walls on imaging studies is a good diagnostic clue for this rare disease. For optimal treatment of venous ACD, complete resection of cystic wall and material is the best method of treatment at present.

#### CONFLICT OF INTEREST

None.

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